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## First Case of Visceral Leishmaniasis Caused by *Leishmania martiniquensis*

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**Abstract.** We report the first case of visceral leishmaniasis (VL) caused by *Leishmania martiniquensis* in the Caribbean, which until now, was known only to cause cutaneous leishmaniasis. The disease presented with fatigue, anemia, and hepatosplenomegaly in a 61-year-old man with human immunodeficiency virus (HIV) infection who was receiving antiretroviral therapy. Diagnosis was made by bone marrow biopsy. VL is life-threatening, and its emergence in the Caribbean is of concern.

### INTRODUCTION

Intracellular protozoan of the *Leishmania* genus, mainly transmitted by sandflies, are the causative agents of leishmaniasis. More than 12 million people are currently infected worldwide.<sup>1</sup> Among its different clinical presentations, visceral leishmaniasis (VL) is life-threatening. Human immunodeficiency virus (HIV) infection is one of the major risk factor for developing VL and reported in 2–12% of all cases.<sup>1,2</sup>

The new autochthonous and divergent *L. (L.) martiniquensis* n. sp. was first isolated in 1995; its taxonomical position was established in 2002, and it was named in 2014.<sup>3–5</sup> This species is up to now restricted to Martinique and has been only reported in patients with cutaneous lesions. We report the first case of VL caused by this parasite in an immunocompromised HIV-infected patient.

### CASE REPORT

A 61-year-old heterosexual Caribbean male was diagnosed with acquired immunodeficiency syndrome (AIDS) prurigo in 2006.<sup>6</sup> He worked as a painter and a coconut picker and seller. He was born and had always lived in Martinique, except from 1994 to 2001, when he had lived in Guadeloupe. He had traveled to northern Europe and Haiti. His sole medical history was hypertension. He had never used intravenous drugs. At the time of HIV diagnosis, no opportunistic infection was found. Immunological, virological, and therapeutic data are summarized in Table 1.

Combination antiretroviral therapy (cART) was introduced in May of 2007, with significant reduction of HIV viral load and increased CD4+ cell count 1 month later. However, at 1 year, although there was clinical improvement of prurigo and continued viral suppression, CD4 counts were decreasing. Genotypic testing for HIV-1 drug resistance on an initial sample had shown no transmitted resistance, and drug monitoring revealed normal absorption. Despite cART regimen switching

in January of 2010, the CD4 counts continued to decline, and the patient remained virally suppressed (Table 1).

He progressively developed hepatosplenomegaly and a normochromic normocytic aregenerative anemia of < 10 g/dL. The patient had no fever but reported permanent fatigue. Sulfamethoxazole/trimethoprim (SMX-TMP) toxicity was hypothesized because of a reduced serum folate level, but folic acid supplementation failed to correct it. A bone marrow biopsy performed in November of 2011 revealed intrahistiocytic parasites consistent with the amastigote forms of *Leishmania* spp. (Figure 1).

The diagnosis was confirmed by *Leishmania* polymerase chain reaction (PCR) realized on whole blood with RV1/RV2 probes targeting a kinetoplastic DNA locus (145 bp).<sup>7</sup> The molecular identification based on the ribosomal 18S RNA locus analysis gave a 100% identity with the sequence of the MHOM/MQ/92/MAR1 strain (GenBank accession number AF303938.1), a divergent *Leishmania* strain described for the first time in Martinique in 1995 and recently named *L. martiniquensis*.<sup>4,5</sup> A retrospective analysis of several sera from our patient stored since 2007 detected high levels of antileishmanial antibodies by indirect immunofluorescence assay (IFA) and enzyme-linked immunosorbent assay (ELISA) using *L. infantum* antigen (Table 1). Immunoblot revealed two specific bands of 14 and 16 kDa, confirming the specificity of these antibodies.

The patient was treated with liposomal amphotericin B (4 mg/kg equal to 300 mg/day from day 1 to 5 and on days 10, 17, 24, 31, and 38 for a cumulative dose of 3 g) and also required a blood transfusion. Symptoms rapidly improved, with decreasing hepatosplenomegaly and disappearance of fatigue, and the patient remained asymptomatic more than 1 year later. Significant increases of hemoglobin level and CD4+ cell count above 350/mm<sup>3</sup> followed as well as a negative blood *Leishmania* PCR 20 months later. The patient did not receive secondary prophylaxis.

### DISCUSSION

Leishmaniasis is endemic in Central and South America but rarely occurs in the Caribbean.<sup>8</sup>

The first cases of presumed autochthonous cutaneous leishmaniasis (CL) in Martinique were diagnosed based on direct

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species in the Caribbean. *Lutzomyia atroclavata* has been identified in Martinique, Guadeloupe, and the Virgin Islands; black rats (*Rattus rattus*), mongooses (*Herpestes auro-punctatus*), marsupials (*Didelphis marsupialis*), and canids are all potential animal reservoirs that should be investigated.<sup>8,16</sup>

The emergence of *L. martiniquensis* infection with the possibility of visceral extension could be of concern in the Caribbean region, where the prevalence of HIV infection is high.

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